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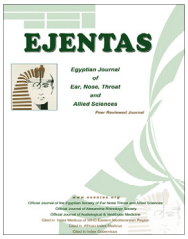

REVIEW

Dysphagia and dyspnea by lingual thyroid mass: An appropriate approach

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Abstract Lingual thyroid is a rare embryological anomaly originated from the thyroid gland failure that descends from the foramen cecum to its normal eutopic pre-laryngeal site. The case in this study was a 39 year old female, presenting with the sensation of a foreign body, progressive dysphagia and dyspnea. Indirect laryngoscopy revealed a large well-defined mass in the tongue base. Imaging studies confirmed the diagnosis of large ectopic lingual thyroid. The surgery was performed via an external cervical approach due to the mass size. The decision on the best treatment looks into the mass position, size, symptoms, airway emergency and medical facilities.

Contents

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1. Introduction

Lingual thyroid is a rare developmental anomaly that originates from aberrant embryogenesis during the passage of the thyroid gland through the neck. Embryonic development begins around 24 days after fertilization in the floor of the Primitive hypopharynx out of the median endodermal thickening. The primitive gland starts to descend close to the hyoid bone and the laryngeal cartilages.[1]

The thyroglossal duct is a narrow tube which connects the developing thyroid gland to the tongue and it usually involutes during weeks 6–8. The foramen cecum is the opening of the thyroglossal duct in the tongue. The thyroid gland gradually descends to meet the lateral ultimo-branchial bodies; the fusion of these elements forms the functional and mature thyroid gland by the third fetal month.[2]

Lingual thyroid is the most frequent ectopic location for the thyroid gland, although its prevalence varies between 1:100,000 and 1:300,000. The clinical incidence is reported to range from 1:4000 to 1:10,000. Its true incidence is probably underestimated; some authors reported the ectopic lingual tissue 10% out of 200 consecutive necropsies.[3] In literature, approximately 400 symptomatic cases were presented.[4] Ectopic thyroid tissue can also occur between the geniohyoid and mylohyoid muscles (sublingual thyroid), above the hyoid bone (pre-laryngeal thyroid) or in such rare sites as the mediastinum, precardial sac, heart, breasts, pharynx, esophagus, trachea, lungs, duodenum, mesentery of the small intestine, and adrenal gland.[5–9]

Most ectopic thyroids are asymptomatic; instead, patients complain from some upper airway problems such as foreign body sensation, progressive dysphagia to solid foods, odynophagia, hoarseness, hot potato voice, bleeding and dyspnea. Some patients have difficulty in breathing or snoring on their back position. Although symptoms grow very gradually, some patients may experience acute onset of obstructive airway symptoms.

2. Case

A 39-year-old female was admitted with dyspnea and muffle voice by the emergency ward at Imam Reza Hospital, Tabriz, Iran. The case had a mass in the tongue base with a foreign body sensation and stomatolalia that caused dyspnea and dysphagia to solid food. The patient was unable to lie on her back for last 5 years. The airway obstruction caused mouth breathing and snoring. These symptoms showed gradual growing in the last 10 years. Weight loss was another unexpected symptom. No record for thyroid disease was available except an untreated mild depression.

In the oral examination, the patient had a solid, pink and spherical mass, covered with intact mucosa on the base of the tongue, obstructing the visualization of the larynx. Neck examination revealed neither palpable thyroid glands in the normal pre-tracheal position, nor cervical adenopathies. The computed tomography (CT) revealed a $41 \times 34 \times 45$ mm mass with distinct margins restricted to the tongue base enhancing after contrast administration. Absence of the thyroid glands was notable (Fig. 1).

A thyroid scan with technetium Tc-99m sodium showed a marked isotope uptake in the area of the tongue base with no uptake in the neck (Fig. 2). Tests of thyroid hormones showed normal thyroid-stimulating hormone (TSH) concentrations and normal Free T3 and Free T4.

3. Surgical technique

In this case, an external approach with transhyoid median pharyngotomy was utilized. The anesthesiologist was unable to intubate the patient even with glidescope due to the large mass kissing the posterior pharynx wall. Although awake fiberoptic nasotracheal intubation is reported as the common option in these patients, the tracheotomy under local anesthesia was initially done due to the insufficient equipment. It was followed by the operation under general anesthesia.

An 8 cm transverse midline incision was done in the neck under the hyoid bone. The hyoid bone and the suprahyoid muscles were exposed, the infra-hyoid and supra-hyoid muscles were freed and the hyoid bone body was removed. Another incision was done in the pharyngeal mucosa to enter the hypopharynx. The mass visualized at the tongue base was dissected with an under mucosa approach and excised (Figs. 3 and 4). The pharyngeal opening was sutured in 2 layers, and the neck incision was closed in 3 layers. A nasogastric tube was introduced to feed the patient for 10 days. The tracheostomy tube was removed after 5 days. On the 8th post-operative day, the patient was discharged. The substitutive hormone therapy was commenced to maintain the euthyroid state. The excised specimen was sent for microscopic examination which was reported as a normal thyroid tissue.

4. Discussion

A rare embryological aberration, ectopic thyroid may occur when the migration of the thyroid results in the lingual (at tongue base), sublingual (below the tongue), prelaryngeal (in front of the larynx), and substernal (in the mediastinum) ectopy. Dual ectopic thyroid is described[1,10] even with the thyroid glands in the normal location.[11] It is more frequent in females with the ratio 4:1 to men. Ectopic thyroid is seen at any age but more commonly during childhood, adolescence or around menopause. This anomaly probably occurs when the demand

Figure 1 CTscan with contrast revealed a large enhanced mass at tongue base.

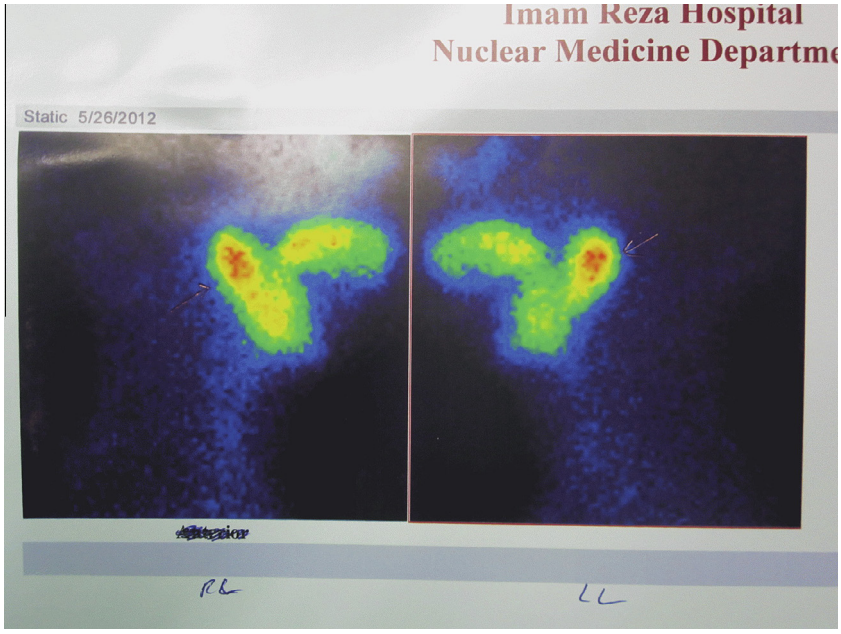

Figure 2 Scan with Tc-99m showed massive uptake at tongue base but no uptake at neck.

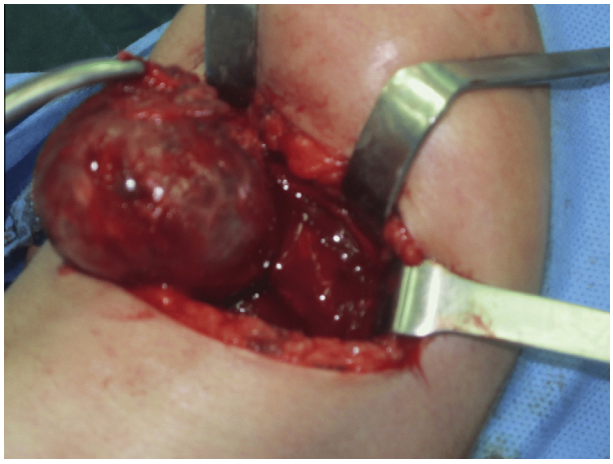

Figure 3 The mass was dissected within pharynx through transhyoid approach.

for thyroid hormones increases while it will increase the circulating TSH levels with the growth of ectopic thyroid tissues.[12]

About 33–62% of the patients with ectopic thyroid show hypothyroidism with increased level of TSH.[5,13] Most ectopic thyroids are asymptomatic with no required therapies. Symptoms are related to the growth of the thyroid tissue, causing dysphagia, dysphonia, stomatolalia, bleeding or dyspnea.[4–14] Acute airway compromise in these patients is very rare in literature, although it is possible because of acute bleeding in the ectopic thyroid. Management in this emergency situation depends on available facilities. Fiberoptic-assisted intubation seems the best but temporary tracheostomy and positive pressure mask ventilation are other alternatives. The enlarged lingual thyroid has been described as a cause of difficult intubation during induction of anesthesia, especially in case of bleeding.[10,14]

Clinically, lingual thyroid presents itself as a pink and firm mass at the tongue base. The most important diagnostic tool is the thyroid scan with technetium Tc-99m sodium. Ultrasonography, CT and magnetic resonance imaging (MRI), however, may help to define the extension and location of the ectopic thyroid gland. Thyroid scan can also reveal whether there are other sites of thyroid tissues. In approximately 75% of the patients, the ectopic tissue is the only functioning thyroid tissue in the body.[15] Therefore, it is important to follow up the patients for the risk of post-operative hypothyroidism.

Differential diagnosis for tongue base mass includes lymphangioma, minor salivary gland tumors, midline bronchial cysts, thyroglossal duct cysts, epidermal and sebaceous cysts, angioma, adenoma, fibroma and lipomas.[5,10]

Managing lingual thyroid is still controversial. There is no consensus about the optimal therapeutic strategy, perhaps due

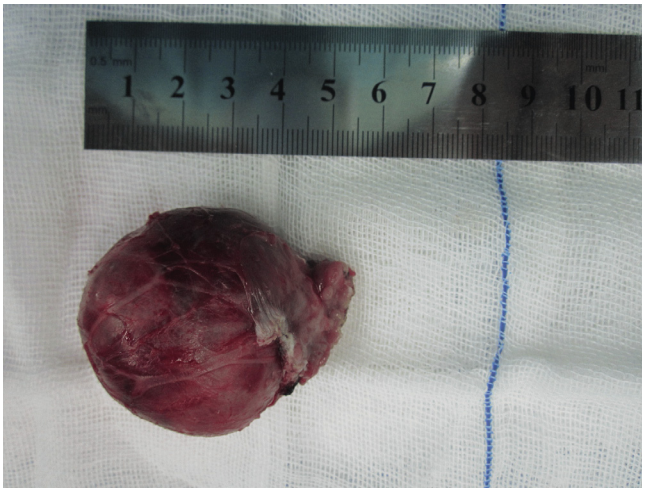

Figure 4 Large well defined thyroid mass was removed.

to the rarity of this clinical entity.[16] No treatment is required when the lingual thyroid is asymptomatic and the patient is in the euthyroid state. Careful observations are recommended in order to trace any developing complications. Malignant transformation has been reported in a few cases.[17]

Some researchers consider the complete surgical removal of the thyroid gland as an appropriate treatment.[18–21] For patients with no or mild clinical symptoms and elevated TSH concentration, the substitutive therapies with thyroid hormone might be successful to gradually reduce the mass size. Ablative radioiodine therapy is another alternative approach recommended in aged patients or patients unfit for surgery. This treatment should be avoided in children and young adults as the required systemic dosage has some potentially damaging effects on the gonads or other organs. Moreover, the thyroid tissues are often hypoactive and the required dosage of radioiodine is generally high.[22]

Surgery is the best option when medical therapies fail in symptomatic or complicated cases. In literature, several surgical approaches are described such as surgical removal of the mass with external approaches, trans-hyoid, lateral pharyngotomy, or trans-oral ablation of the mass. The trans-oral approaches seem to prevent injuries to deep neck structures and other possible complications (injuries to the lingual nerve, fistula formation, deep cervical infection and visible scars) and could be considered as a significant treatment for small masses. Since the external approaches with temporary tracheotomy prevent bleeding, they are recommended in the case of bulky masses.

For trans-oral approaches, cold instruments with monopolar coagulation and laser CO_2 are known as practical.[14,23] New technique has been recently used for surgery is Transoral Robotic Surgery.[24] Although authors suggested this method as a feasible and safe alternative to other invasive surgical approaches in patients with symptomatic lingual thyroid, but experience with this technique is so low and the equipment may be unavailable anywhere.

The possibility that the ectopic thyroid tissues could be the only functioning tissues must be carefully considered. By some researchers, transplantation of the excised ectopic tissue is recommended to avoid permanent hypothyroidism.[4] As an alternative, substitutive hormone treatment may start to preserve the euthyroid states. Transplantation is not necessary in the case of partial surgical eradication since the substitutive therapy seems necessary to preserve euthyroid state and avoid recurrence of the mass.

Based on the results in this case study, the researchers believe that the choice of the appropriate treatment is somehow problematic considering patients' general conditions, the lesion size and the presence of local symptoms or complications such as hemorrhage, cystic degeneration or malignancies.[18–21] In symptomatic, old patients, unfit to undergo more aggressive surgical approaches, or in small and anterior lesions, the partial trans-oral approaches are recommended. The partial trans-oral approach is less aggressive but cannot prevent malignant transformation and possible relapses. Moreover, it allows less control of bleeding during the surgery.

Total ablation is considered as the most appropriate approach in younger patients, in the case of large lesions or deeply located in the caudal part of the tongue base. In such cases, transplantation of the thyroid tissue is not necessary since substitutive hormone treatment is highly recommended as a radical surgical approach for normal thyroid gland.[25]

Special care should be taken to ensure the patient's airway patency before and after surgery. The major risk to these patients is that insertion of any object into the oral cavity could traumatize the glandular tissue causing subsequent hemorrhage and a compromised airway. This could occur with a simple oropharyngeal airway or during laryngoscopy. Some authors suggested procedures requiring general anesthesia in these patients should be carried out using fiberoptic-assisted nasal intubation.[26] Temporary tracheostomy is another choice if fiberoptic-assisted intubation was not available as in our case, she underwent temporary tracheostomy for 5 days. After surgery also maintaining the airway is critical and it can usually be achieved by temporary tracheostomy or maintaining the naso-tracheal tube for the first hours after surgery. Tracheostomy or intubation should not be removed until hemostasis and airway patency have been confirmed.[25]

5. Conclusion

The critical point in diagnosing lingual thyroid is considering this entity. In the case of patients complaining from spherical masses, conducting indirect laryngoscopy and observing the tongue base may raise clinical suspicion while the accessible imaging options may confirm diagnosis. The patients are often admitted in the emergency ward with a sudden increase in the mass size, asthma and dyspnea; hence, the lingual thyroid is a possibility.

Most researchers believe that the best treatment is determined by the mass position, size, symptoms, airway emergency, medical facilities such as laser therapy, and last but not the least the experience of the surgeon. The operated patients need a follow-up treatment for side effects or hormone substitution.

Conflict of interest

None.

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